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Case report

Incidental bilateral xanthogranuloma of the lateral ventricles at autopsy — A case report



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ABSTRACT

We report the case of a woman found dead in a public garden. She had multiple contusions, bruises and lacerations of the face due to blunt force trauma. Microscopic examination of the brain was consistent with a death due to severe craniocerebral injury. The neuropathologist also found yellow nodular lesions located in each lateral ventricle and composed by cholesterol clefts, foamy macrophages, and giant cells. A diagnosis of xanthogranuloma of the choroid plexus was made. According to the literature, xanthogranulomas are uncommon benign lesions of the central nervous system. Mostly are found incidentally at post-mortem examination. However, some of them are symptomatic and can lead to severe nervous central damages if they remain untreated.

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1. Introduction

Xanthogranulomas, also known as cholesterol granulomas, are tissue reactions with a pseudo-tumor aspect, composed of cholesterol clefts, inflammatory cells such as foamy cells, histiocytes, multinucleated foreign body giant cells, hemosiderin deposits, and fibrous proliferation. The etiology is still a matter of debate. Xanthogranulomas are benign lesions, which rarely involve the central nervous system. They are mostly located in the choroid plexus of the lateral ventricles, and are idiopathic, and usually asymptomatic, with an autopsy incidence of about 1.6–7.0%. The majority of symptomatic cases concern the choroid plexus of the third ventricle, and are idiopathic, and usually asymptomatic, with an autopsy incidence of about 1.6–7.0%. The majority of symptomatic cases concern the choroid plexus of the third ventricle, as of bilateral xanthogranuloma of the choroid plexus of the lateral ventricles, found incidentally at autopsy.

2. Case report

A 50-year-old female was found dead in a public garden. At external examination, she had multiple lacerations of the head and a large, horizontal slash wound of the neck. At autopsy we noted

multiple fractures of the face and skull, a left temporo-parietal and a right frontal subdural hematoma, and a diffuse, bilateral subarachnoid hemorrhage. There was no injury of the respiratory tract and no vascular injury associated with the slash wound of the anterior part of the neck. Toxicology showed the presence of tiapride (an antipsychotic), and meprobamate, both at infratherapeutic level, associated with a moderate blood alcohol concentration (0.044%). Neuropathology showed cortical contusions, a cerebral edema and a mild subarachnoid hemorrhage of the brain. These findings were consistent with blunt force trauma. Moreover, one nodular lesion of the choroid plexus was found in each lateral ventricle (Fig. 1). It was composed of foamy macrophages heaps, lymphoid cells, multiple deposits of cholesterol clefts surrounded by multinucleated giant cells, and basophil rounded calcifications (Fig. 2). A diagnosis of xanthogranuloma of the choroid plexus was made. Death was attributed to blunt force trauma and was ruled a homicide. The xanthogranulomas were not considered as a contributing factor.

3. Discussion

Intra-cranial xanthogranulomas are uncommon. Aside from the choroid plexus, some authors reported cases of xanthogranulomas in various locations, such as the dura mater,⁵ and the supra-sellar region where it can easily be misdiagnosed as a craniopharyngioma.⁶ The first description of a xanthogranuloma was reported

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Fig. 1. A: Vertical brain sections of occipital lobes, B—C: Yellow nodular lesions of the choroid plexus in lateral ventricles. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

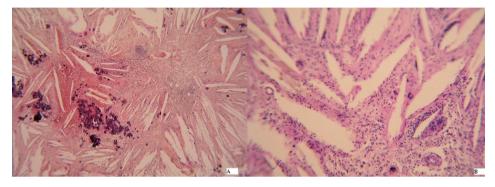


Fig. 2. A: Nodular lesions of the choroid plexus. Multiple prints of cholesterol clefts with inflammatory reaction of foamy macrophages and multinuclear giant cells (\times 40). B: Cholesterol clefts and basophils calcifications (\times 160).

by Blumer in 1900. The pathogenesis is not fully understood, the etiology being whether neoplastic, inflammatory or metabolic. However, we found in the literature a preponderant theory, proposed by Shuangshoti et al.⁷ to explain the development of these lesions within the choroid plexus. Desquamated epithelial cells enter the interstitium of the choroid plexus via a disruption of the basal lamina, release its lipid content, which accumulates as these cells degenerate, and then the lipids lead to a xanthogranulomatous reaction with an accumulation of inflammatory cells.^{4,5,7–9} Thus, some authors believe that a neuroepithelial cyst is formed at the origin, which evolves either to a colloid cyst, to a xanthogranuloma or to an intermediate form.³

In our case, a typical xanthogranuloma was found. As reported in previous cases, xanthogranulomas are most likely smooth, pale yellow masses, 3,6,8–10 and occasionally encapsulated. Microscopically, these tumors are composed of cholesterol clefts, hemosiderin deposits chronic inflammatory cellular reaction with foamy cells, macrophages, and multinucleated giant cells. 4–6,8–10 Secondary changes can occur such as intralesional hemorrhage, fibrosis, or focal calcifications. It explains the absence of an epithelial covering, which distinguishes them from colloid cysts.

Computed Tomography-density of xanthogranulomas varies from hypo- to hyperdense in comparison to brain tissue. On Magnetic Resonance Imaging (MRI), they usually appear isointense or hypointense on T1-weighted images, hypointense on T2-weighted images, and a variable enhancement following gadolinium administration. ^{6,9} Yet, the radiological features are very inconsistent, due to the heterogeneity of their content. ¹⁰ The presence of focal calcifications is reflected by a hyperintensity on T2-weighted images. ^{3,5} Therefore, the differential diagnosis with other masses of the lateral ventricle, such as meningioma, papilloma, ependyma, arteriovenous malformations, ⁹ remains difficult.

Our victim did not have any medical follow-up, so the asymptomatic characteristic or otherwise of her condition is questionable. Xanthogranulomas are usually found within the choroid plexus of the lateral ventricles, being benign and rarely symptomatic, and are

discovered incidentally. They can sometimes be revealed by visual disturbance, due to a peripheral edematous infiltration.³ In contrast, when they are located in the third ventricle, they can be symptomatic by obstructing the cerebrospinal fluid flow causing obstructive hydrocephalus, or even be the cause of sudden death by herniation or massive hematomas causing hypothalamus inhibition.¹⁰ P. Miranda et al. described a case of a symptomatic xanthogranuloma in the region of the third ventricle and hypothalamic area, associated with edematous infiltration in the basal ganglia of the left cerebral hemisphere, being responsible for gait and urinary disturbances, and cognitive impairment.³ Kent J. Donelan et al. reported a sudden death of a 51-year-old-man due to an acute subarachnoid and intraventricular hemorrhage secondary to a hemorrhage within a choroid plexus xanthogranuloma of the left lateral ventricle. The post-operative course of a total excision of a symptomatic xanthogranuloma often showed a partial or total normalization of the clinical state^{3,5,8,10} and in a few cases remained uneventful.9

Although xanthogranulomas are benign lesions, they can result in disabling symptoms^{3,5,6,8–10} or even disastrous consequences if left untreated.⁴ The forensic pathologist needs to balance both sides to ensure that xanthogranulomas do not have a direct, or even an indirect link with the death. Therefore, these lesions should not be underestimated. In living individuals, xanthogranulomas should be followed up if asymptomatic and be an indication for surgery according to its location, its MRI aspect and its symptoms.

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Conflict of interest

The authors declare no conflict of interest.

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